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CASE REPORT

An uncommon cause of unilateral hand and forearm oedema

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Abstract A case with an uncommon cause of unilateral hand and forearm oedema in a 37-year-old man is presented. Complex regional pain syndrome type I was diagnosed and treated with mannitol infusion, dimethyl sulfoxide, vitamin C, ketanserin and carnitine. Also, physiotherapy and occupational therapy were started. None of these treatment modalities resulted in reduction or prevention of recurrence of the oedema. All diagnostic procedures did not reveal a cause for this oedema.

Keywords Hand · Forearm · Oedema · Uncommon

Introduction

The term "oedema" is used in the case of excess volume of fluid in the extracellular–extravascular compartment (the interstitial space). Most commonly, the fluid accumulates in the dependent parts of the body due to the effects of gravity. Oedema can be generalised or localised, and this classification can be a helpful guide when investigating its cause [3].

Oedema of the hand and forearm can have different causes. Oedema after contusion or a fracture is well known. During an infection or after burn injury, it can also be expected. Mostly, it is a gradual process and will resolve during different therapies.

Swelling—pitting or non-pitting oedema—is usually present when complex regional pain syndrome type I (CRPS I) is diagnosed and localised to the painful region. One of the clinical features in these cases is a more diffuse swelling (generalised oedema). Uniform diagnostic criterion for CRPS I is (consensus conference IASP 1995) the presence of four out of the following five symptoms: unexplained diffuse pain, color changes relative to the contralateral extremity, temperature changes relative to the contralateral extremity, diffuse oedema or limitations in range of motion [6]. When CRPS I is diagnosed, symptoms occur or are aggravated during or after exercise. Also, symptoms are present over a larger area and distal to the primary site of injury or trauma.

Localised oedema is limited to a particular organ or vascular bed. Unilateral extremity oedema is usually due to venous or lymphatic obstruction, such as deep venous thrombosis, tumour obstruction or post-coronary bypass with a harvested leg vein [5]. Stasis oedema of a paralysed lower or upper extremity may also occur. A more balloon-like aspect of oedema is seen after, for example, a constricting bandage (localised oedema) [3].

Oedema delays healing and causes pain and stiffness and thereby compromises functional healing of an extremity. It is a constant challenge for hand surgeons and hand therapists to prevent oedema. During treatment of oedema, the principles of elevation and active motion are therefore important. Each cause of oedema has its own specific

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Fig. 1 Unilateral swelling of the patient's right hand and forearm

treatment modality. We present a case with an unknown cause of unilateral hand and forearm oedema in a 37-year-old male patient.

Case report

A 37-year-old man sustained a fall down the stairs causing a slight contusion to his right non-dominant forearm and hand. There were no fractures, and no acute oedema was present. There was no previous history of trauma to his right arm. The patient was advised to elevate and rest his hand for some days.

Several weeks later, the patient returned to the hospital with a hand and forearm oedema without an obvious cause. CRPS I was diagnosed. Treatment was started with mannitol infusion [1], dimethyl sulfoxide [2], vitamin C [10], ketanserin and carnitine [7] and physical and occupational therapy.

The swelling decreased during hospital stay, and the patient returned home. A few days later, he returned to the hospital because of progression of the swelling of his hand and forearm. Magnetic resonance imaging (MRI) and ultrasound did not show a cause for the diffuse swelling. Also, a ganglion block did not decrease the swelling.

The patient was sent to the department of rehabilitation in our hospital for a second opinion. A clinical evaluation showed unilateral swelling of his right hand and forearm (Figs. 1 and 2). Initially, the patient refused inspection of his right upper arm because he had no complaints in that region. Because of the progression of the swelling and his refusal to allow inspection of his arm, automutilation was considered as a possible cause of the swelling. Also, strangulation signs were seen on his upper arm (Figs. 1 and 2). Doppler studies and new laboratory tests did not exclude the possibility of automutilation. A lymphoscintigraphy did not indicate a late onset lymphoedema. The patient, however, denied any form of automutilation. Psychological help was arranged. Because the patient consistently denied any form of automutilation, it was difficult for the psychologist to treat him. During the hospital stay, the swelling decreased amazingly quickly within 10 days, with the only treatment modalities being

Fig. 2 Strangulation signs seen on the patient's upper arm

observation and immobilization with a plaster of paris splint. In the meantime, fixed contractures of all his fingers were observed. The patient had an end stage of an afunctional hand and forearm. During follow-up consultations, oedema recurred to a minor degree, but the patient is, after 24 months, still unable to work and has been on sickness benefit since the original injury. The request for amputation by the original treatment centre and the patient was not followed up.

Discussion

The classification of oedema into generalised or localised can be a helpful guide in investigating its cause. Generalised oedema is soft tissue swelling of most or all regions of the body. Localised oedema is limited to a particular organ or vascular bed. Oedema delays healing and causes pain and stiffness and thereby compromises functional healing of an extremity.

In the first instance, the frequent occurrence of swelling after a trauma was suspected, but because of the steady progression of the swelling, other possible causes were considered. No fractures were seen on X-ray. MRI or ultrasound showed no obvious cause such as thrombosis or tumour. Blood tests did not reveal an infection or rheumatic disorder. Other causes such as lymphogenic pathology were excluded because of a normal lymph scintigraphy.

CRPS I was a possible diagnosis, but during clinical evaluation, the possibility of automutilation was discussed. Our patient had no psychiatric history. Because of the very fast progression of oedema in our case and also the balloon-like appearance, we considered automutilation as a most likely cause. Localisation on one side, fast progression of oedema out of hospital and fast regression during hospital stay are factors that make automutilation a distinct possibility. Signs of strangulation on his upper arm confirmed our suspicion. Most frequently, automutilation occurs (in about one third of all cases) in patients with psychiatric disorders. Potential patients are difficult to recognize [4].

Thirty years ago, Smith [8] published a large series of patients with factitious lymphoedema of the hand, sometimes caused by a tourniquet and proximally ending in a circumferential discoloured constriction ring.

The findings of strangulation of the affected upper extremity oriented to the diagnosis of a factitious disorder or Munchausen's syndrome. In this psychiatric condition, the patient simulates a clinical condition in a very convincing way, mimicking to perfection a disease. Usually, the patients are either crying for attention or causing some sort of automutilation as a means of punishment by unconscious guilt. Mostly, the sole purpose is assuming the sick role. It is important to always keep in mind that patients with unexplained and unspecific signs, such as oedema present in this case, may be indications of Munchausen's syndrome. After identification of this syndrome, the patient should be referred to a psychiatrist.

Of course, in our patient, automutilation in combination with CRPS I could be a possibility. The features in our patient also resemble the clenched fist syndrome [9]. The development of a swollen clenched fist is thought to be "a symbolic expression of forbidden wishes". It "represents feelings of anger that are suppressed by paralyzing the hand". Swift reported that resistance to treatment is a feature of this syndrome. Although our patient did not refuse therapy, further psychological intervention was cancelled, and he was made aware of his own responsibility in preventing a recurrence by the psychologist involved.

In conclusion, oedema is a very simple clinical sign but can reveal serious diseases. Our case report represents an unusual cause of unilateral oedema of the hand and forearm. Localisation on one side, fast progression out of hospital and fast regression during hospital admissions are factors that make automutilation highly likely. The complexity of these problems requires long-term multidisciplinary therapy.

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